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Case report

# Testicular carcinoid tumor: A case report and literature review

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ABSTRACT

We present a case of a 46-year-old man with an incidental finding of a right testicular tumor on magnetic resonance imaging. He received radical orchiectomy, and the pathological report showed a testicular carcinoid tumor. He has remained in a stable condition 3 years after the operation. Carcinoid tumors of the testis are extremely rare and account for less than 1% of testicular tumors and carcinoids. A painless mass or prominent testis enlargement is the most distinctive presentation. Less than 1–3% of patients demonstrate carcinoid syndrome. Nuclear scintigraphy scan and computed tomography scan have a diagnostic value in excluding carcinoid from other sites. Radical orchiectomy is the treatment of choice, and the prognosis is good for localized carcinoid tumors after resection. Adjuvant chemotherapy or radiotherapy for advanced disease shows only minimal benefits.


## 1. Introduction

Carcinoid tumors, which most frequently occur in the gastrointestinal tract, are uncommon. In a report from the Surveillance, Epidemiology, and End Results (SEER) program of the National Cancer Institute database on 11,427 carcinoid patients treated from 1973 to 1997, the majority of carcinoids were located in the gastrointestinal tract (55%) and bronchopulmonary system (30%).[1] DeVita[2] reported that testicular carcinoids account for less than 1% of all testicular tumors and for only 0.2% of all carcinoid tumors.[3] Embryologically, carcinoid tumors may be derived from foregut, midgut, or hindgut, and the testicular carcinoid tumor has a midgut origin. Testicular carcinoid tumors are composed of uniform polygonal cells with fine, granular chromatin; small nucleoli; and scant to moderate amount of cytoplasm.[4] Herein, we present a case of a primary testicular carcinoid tumor along with a literature review.

## 2. Case report

A 46-year-old man presented to our clinic due to a 2 cm right testicular mass detected incidentally by magnetic resonance imaging (MRI) during a health examination (Fig. 1A and B). The tumor was nontender, ill defined, and firm in consistency on palpation; however, the size of his right testis was normal and measured about 3 cm × 3 cm × 2 cm. No inguinal lymphadenopathy was observed. The patient denied fever or any recent travel. He also denied diarrhea, abdominal cramps, nausea, vomiting, hot flushes, or dyspnea. His hemogram and biochemistry tests were all normal. Tumor marker tests showed the following: alpha-fetoprotein: 4.49 ng/mL (normal 0.00–20.00 ng/mL); beta-human chorionic gonadotropin: 0.08 mIU/mL (normal <5 mIU/mL); and lactate dehydrogenase: 197 U/L (normal 131–250 U/L). Scrotal sonography revealed a heterogeneous echoic nodule 1.9 cm in size in the right testis with central calcification and increased vascularity (Fig. 1C and D). Right radical orchiectomy was performed uneventfully.

Grossly, the tumor was yellowish white, firm, and 1.9 cm × 1.5 cm × 1.2 cm in size. It was confined to the testis, and the tunica albuginea and epididymis were free of the tumor (Fig. 2). Microscopic examination revealed that the tumor was composed of uniform cells bearing round nuclei and growing in a festoon- or ribbon-like arrangement. Fine cytoplasmic granules were seen. Tumor cells were positive for chromogranin, CD56, and synaptophysin immunohistochemically, all of which are markers for neuroendocrine tumors. The tumor was negative for alpha-inhibin (Fig. 3), which excluded other possible germ cell tumors. No other teratomatous component was detected in the specimen. A primary carcinoid tumor was diagnosed. A gastroenterologist was consulted after the diagnosis was confirmed. Upper gastrointestinal tract panendoscopy was performed, and no tumors were detected.

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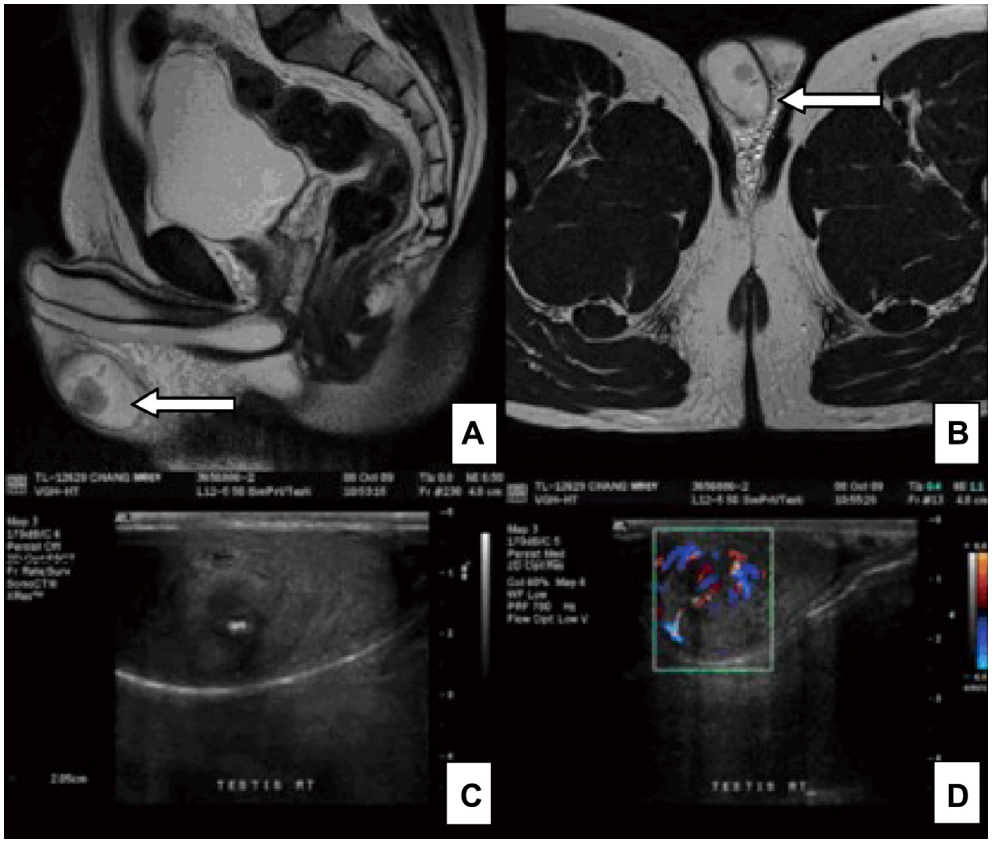


**Fig. 1.** (A and B) Preoperative magnetic resonance image showed a T2 hypointense nodule about 2 cm in size, with coarse calcification being noted in the right testis (arrow). (C and D) Scrotal sonogram showed a heterogeneous echoic nodule 2 cm in size in the right testis with central calcification and increased vascularity.

**Fig. 2.** Yellowish-white, firm tumor, 1.9 cm × 1.5 cm × 1.2 cm in size, in the right testis (arrow).

The patient received follow-up abdominal computed tomography and scrotal sonography regularly, and no evidence of tumor recurrence was found 3 years after the operation. Urine 5-hydroxyindolacetic acid (5-HIAA) level was also checked and found to be within normal limits.

## 3. Discussion

Testicular carcinoid tumors are very rare. They can be divided into three groups: primary testicular carcinoid, carcinoid metastasis to the testis, and carcinoid associated with teratoma. Primary testicular carcinoid is presumed to originate from argentaffin or Kulchitsky cells, or develop from simplified teratoma without other teratomatous elements.[5]

A painless mass or prominent testis enlargement is the two most common clinical presentation of a testicular carcinoid. Less than 1–3% of carcinoid tumors present with carcinoid syndrome, of which carcinoids with metastasis are the most common.[5] A previous study reported carcinoid syndrome in 50% of carcinoids with metastasis and in only 5.6% of nonmetastatic carciniods.[5] Elevation of serum serotonin or urine 5-HIAA levels should raise the suspicion of a metastatic carcinoid tumor.

Because these carcinoids are found mostly in the gastrointestinal tract, especially in the ileum, video capsule endoscopy has been shown to have value in the staging of carcinoid tumors. Somatostatin receptor scintigraphy or iodine-131-metaiodobenzylguanidine scintigraphy may also have a diagnostic value in excluding carcinoids from sites other than the testis.[6] These tests along with computed tomography should be performed to rule out the possibility of other metastatic carcinoid tumors. Somatostatin scintigraphy scan has been demonstrated to be superior than computed tomography scan

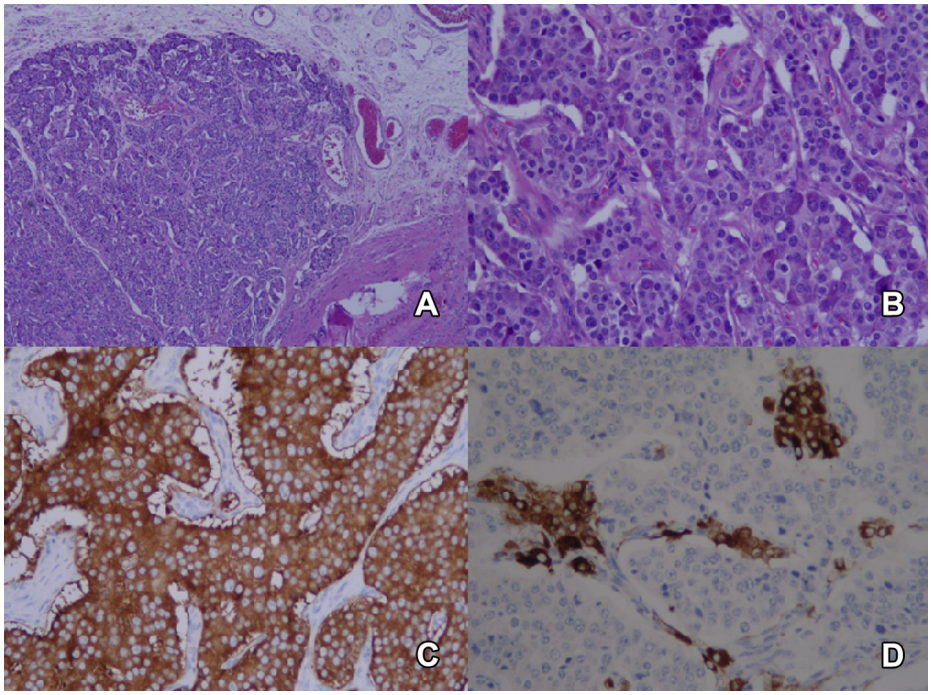


**Fig. 3.** Microscopic findings. Sections show a picture of a carcinoid composed of uniform cells with round nuclei and growing in a festoon- or ribbon-like arrangement under (A) 40× and (B) 100× magnification (hematoxylin and eosin stain). Fine cytoplasmic granules are seen. Under 100× magnification, (C) tumor cells were positive for synaptophysin immunohistochemically and (D) the tumor was negative for alpha-inhibin staining.

in localizing the primary tumor site, with sensitivity up to 96% for detecting metastatsis.[7]

Radical orchiectomy is the treatment of choice for localized testicular carcinoid tumors, and adjuvant therapies including chemotherapy and radiotherapy have been shown to have less optimal effects in the treatment of advanced testicular carcinoids.[6] Moertel and Hanley[8] concluded that chemotherapy for local or metastatic carcinoids provides only limited benefits. Another treatment choice, such as interferon α, has been reported for all carcinoids, with a biochemical response rate of 40%, subjective response rate of 70%, and reduction in tumor size in 20% of patients.[6] Higher dosages did not show obvious benefits in size reduction.[6]

Postoperative follow-up including history taking, physical examination, urine 5-HIAA level, chest X-ray, chest and abdominal computed tomography scans or MRI, and octreotide scintigraphy should be conducted to rule out extratesticular metastasis.[6] Urine 5-HIAA, a metabolite of serotonin, can be measured in a 24-hour urine sample as a marker for follow-up. The sensitivity of this test reaches 75%, with specificity up to 100%.[9] However, the results may be influenced by the ingestion of certain drugs and foods. Plasma chromogranin A is another marker that is useful in detecting neuroendocrine tumors; however, it has higher sensitivity and lower specificity than 5-HIAA.[4] Our patient received whole-body MRI during a health examination and showed no other problems aside from the testicular tumor; there was no evidence of recurrence or metastasis after 3 years of follow-up.

The prognosis of a localized testicular carcinoid tumor after operation is excellent. In an article, Wang et al[4] reported on 29 cases of primary testicular carcinoid tumors of the testis. All the 20 patients with primary testicular typical carcinoid tumor were alive without recurrence. Zavala-Pompa et al[10] reported 57 patients with primary testicular carcinoid tumor: 43 had pure carcinoid tumor and 14 were associated with mature teratoma. Six patients developed distant metastasis, and most of them had larger tumors of pure carcinoid type. However, a long-term follow-up was suggested, as isolated cases of testicular carcinoids have been reported to exhibit an aggressive metastatic course following a prolonged disease-free interval .[4] Our patient received whole-body MRI during a health examination and showed no other problems aside from the testicular tumor; there was no evidence of recurrence or metastasis after 3 years of follow-up.

## Conflicts of interest

The authors declare that they have no financial or non-financial conflicts of interest related to the subject matter or materials discussed in the manuscript.

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